

Conservative management of intra-abdominal haemorrhage associated with angiomyolipoma in the third trimester.

L. McQuillan¹, K. Golding¹, V.Wallis¹, B. Laverick², M. Andrews², S. Holland², A. Khadi³

1. Department of Obstetrics and Gynaecology, Norfolk and Norwich University Hospital Foundation Trust | 2. Maternal Medicine Centre, Norfolk and Norwich University Hospital Foundation Trust | 3. Department of Urology, Norfolk and Norwich University Hospital Foundation Trust

Background

Angiomyolipomas (AML) are benign tumours presenting most commonly in the renal parenchyma and composed of poorly differentiated adipose, smooth muscle and vascular endothelial tissues ⁽¹⁾. They are highly vascular in nature and have a prevalence of 0.2% in males and 0.29% in females with overall prevalence increasing with age. Angiomyolipoma diagnoses can be made as incidental findings and are also associated with complications such as rupture with retroperitoneal haemorrhage (Wunderlich’s Syndrome) and declining renal function both associated with increased size of tumour ⁽²⁾.

Angiomyolipoma in pregnancy is a rare presentation which according to the available literature is associated with a high likelihood of preterm birth. A previous study has noted 100% of angiomyolipoma cells expressing oestrogen receptor beta⁽³⁾ and it is by this means that observed higher rates of growth of angiomyolipoma during pregnancy and hence rupture are hypothesised to occur ⁽⁴⁾. In this case we describe an uncomplicated birth at 37 weeks following conservative management.



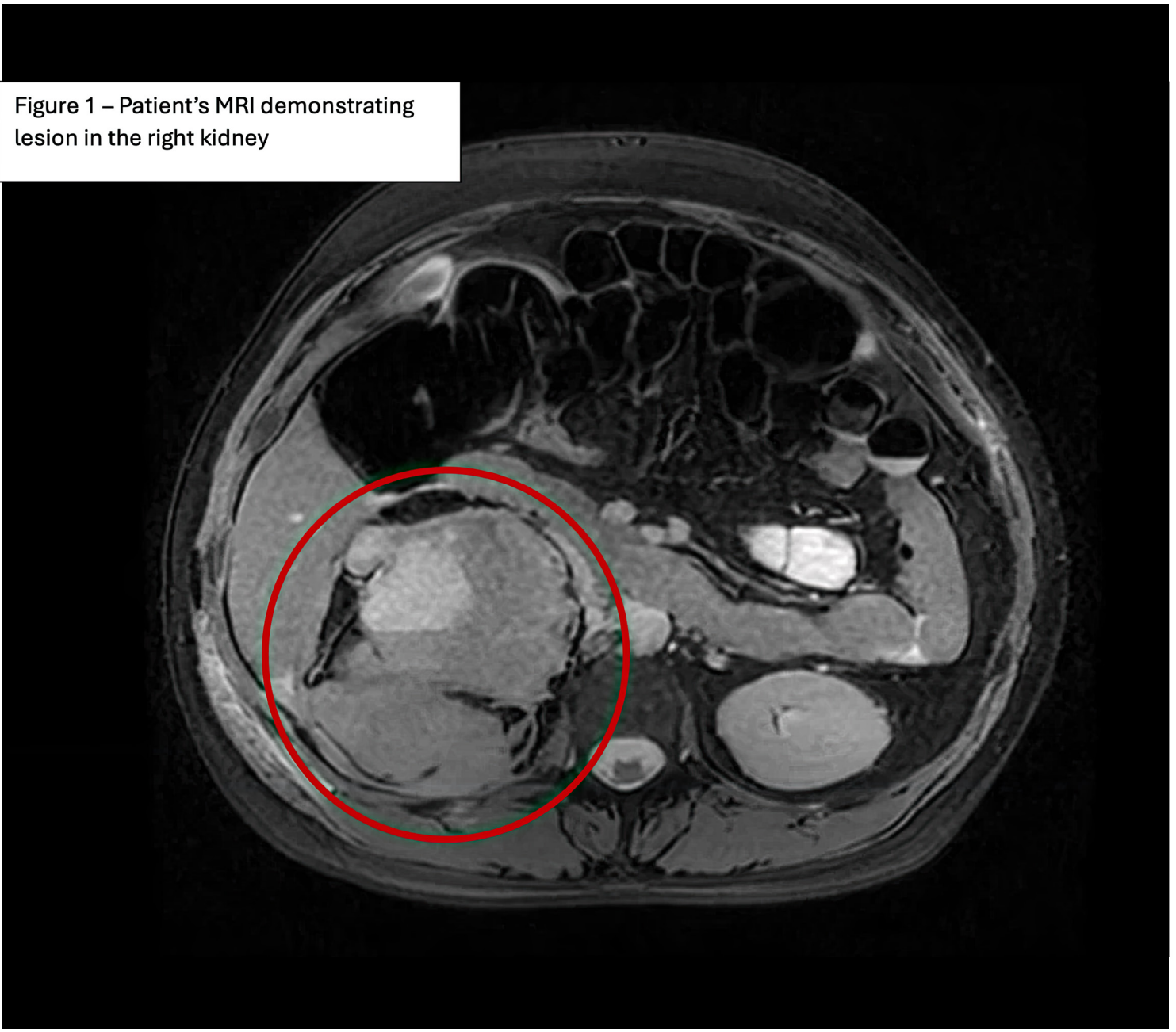
Case report:

A 44 year old with a history of subfertility presented at 27 weeks in her second pregnancy (following previous early miscarriage) with severe loin pain, fever and feeling generally unwell and was treated for pyelonephritis. A renal ultrasound identified a large, haemorrhagic mass suspicious for malignancy. She was transferred to a tertiary centre for urology care and input from the maternal medicine centre was sought with precautionary administration of steroids for foetal lung development.

Of note, she had developed a deep vein thrombosis and pulmonary embolism at 6 weeks gestation in this pregnancy, requiring treatment with low molecular weight heparin. Her past medical history was significant for a previous cardiac ablation in 2023 for supraventricular tachycardia.

An MDT review of MRI imaging suggested a right sided angiomyolipoma with a maximum anteroposterior diameter of 10cm. She developed an acute kidney injury (AKI) and significant anaemia which was managed with blood transfusion and the angiomyolipoma was managed conservatively. She had an uncomplicated caesarean section at 37 weeks with minimal blood loss. She recovered well post-operatively and was discharged on postnatal day 8.

She was followed up in the urology clinic with a repeat MRI and discussion around postnatal surgical management. Subsequent MRI imaging noted a significant reduction in the angiomyolipoma size to 4.6x4.5cm with resolution of the inferior haematoma and the lesion described as well encapsulated. The ongoing management plan was for monitoring with a follow-up scan in 12 months. At time of publication, the patient’s renal function remains somewhat deranged with a creatinine of 90 and an eGFR of 66.



Discussion

This case presented a challenge to both urological and obstetric teams and required inputs from radiology and the haematology also. While in most cases it is theorised that there is tamponade of the concealed haemorrhage ⁽⁵⁾, in this case there was evidence of intraperitoneal leak with a significant Grey Turners sign and ileus which gave complexity to the decision making in taking a conservative approach (as would be the preferred management approach when a patient is haemodynamically stable.)⁽⁶⁾. This was further compounded with the patient’s ongoing requirement for anticoagulation in light of her thromboembolic risk. She also developed gestational diabetes requiring insulin therapy.

The patient required analgesia and intravenous fluids. She had an AKI with a peak creatinine of 127 at 2 days postnatally and developed anaemia (haemoglobin 73g/L) necessitating 5 units of red cell transfusion.

There was a plan for birth by caesarean if there was further bleeding and requirement to transfuse due to risk of foetal compromise. However, with conservative management and close inter-specialty working and monitoring she had an uncomplicated caesarean birth at 37 weeks. If there was evidence of ongoing bleeding there was a plan for radiological embolisation of the lesion at the time of caesarean section. This case provides a rare example of uncomplicated birth at term in a case of AML diagnosed in pregnancy.

Conclusion

This rare, complex case demonstrates the importance of multidisciplinary working to optimise maternal and neonatal outcomes, ensuring all imaging modalities and treatment options are considered. Simultaneously, while planning for all eventualities and managing risk, it is important to avoid overtreating and any unnecessary overmedicalisation of a mother’s birth experience.

References:

1. Bissler, J.J. and Kingswood, J.C. (2004). Renal angiomyolipomata. *Kidney International*, 66(3), pp.924-934.
2. Wang, C., Li, X., Peng, L., Gou, X. and Fan, J. (2018). An update on recent developments in rupture of renal angiomyolipoma. *Medicine*, [online] 97(16), p.e0497. doi:https://doi.org/10.1097/MD.00000000000010497.
3. Boorjian, S.A., Sheinin, Y., Crispen, P.L., Lohse, C.M., Kwon, E.D. and Leibovich, B.C. (2008). Hormone Receptor Expression in Renal Angiomyolipoma: Clinicopathologic Correlation. *Urology*, 72(4), pp.927-932. doi:https://doi.org/10.1016/j.urology.2008.01.067.
4. Kluiving, M.W., Peeters, I., Lely, T.A., Niek van Oorschot and de, L. (2024). The effect of pregnancy on renal angiomyolipoma; a world of knowledge to gain, specifically in women with TSC. *BMC nephrology*, 25(1). doi:https://doi.org/10.1186/s12882-024-03483-4.
5. Rafi, J. and H. Muppala (2009). Retroperitoneal haematomas in obstetrics: literature review. *Archives of Gynecology and Obstetrics*, 281(3), pp.435-441. doi:https://doi.org/10.1007/s00404-009-1282-y.
6. Zapardiel, I., Jesús Delafuente-Valero and José Bajo-Arenas (2011). Renal Angiomyolipoma during Pregnancy: Review of the Literature. *Gynecologic and Obstetric Investigation*, 72(4), pp.217-219. doi:https://doi.org/10.1159/000329328.